Case Report

Adult Intussusception Secondary to Inflammatory Polyps

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Adult intussusception is a rare entity that may present in the acute and subacute setting principally related to the degree of bowel obstruction. Preoperative diagnosis of this condition may be difficult. The intussusception is usually due to a definable intraluminal lesion, most probably neoplasia, unlike intussusception in children. We present the cases of two adult male patients with intussusception. The first presented with acute small-bowel obstruction secondary to a retrograde ileojejunal intussusception with a pseudopolyp as the lead point. This was possibly due to a retrograde ball-valve effect. The intussuscepting segment was resected. The second patient presented with unexplained chronic diarrhoea and an intussusception occurring within the caecum, as demonstrated at colonoscopy, with a terminal ileal pedunculated fibroid polyp as the lead point. A limited right hemicolectomy was performed. Both patients recovered uneventfully and have remained well. A brief literature review of adult intussusception complements the case reports, with an emphasis on the pathogenesis of inflammatory polyps and recommended surgical management. [*Asian J Surg* 2005;28(1):58–61]

Key Words: intussusception, inflammatory fibroid polyp

Introduction

Intussusception is an unusual cause of bowel obstruction in adults.[1] It may present with a variety of either acute or intermittent chronic symptoms that may or may not manifest readily upon clinical examination or routine investigations.[1–3] Retrograde intussusception (distal segment intussuscepting proximally) is not unknown but is rare.[4]

Endoscopic confirmation and, in some instances, therapy for intussusception have been advocated, yet there have been very few reports of endoscopically demonstrable intussusception.[5,6] Alteration in bowel habit as a mode of presentation is also rarely cited.[1–3] We present two contrasting cases of adult intussusception that could provide further insight into this condition.

Case reports

Case 1

A 34-year-old man presented with an unprecedented 2-day history of sudden-onset colicky central abdominal pain associated with bilious vomiting. Clinical examination revealed a dehydrated, relatively thin febrile patient with tachycardia. There was a palpable tender non-expansile central abdominal mass. Bowel sounds were absent.

Routine investigations showed a raised white cell count of 15.8×10^9/L and compensated metabolic acidosis. Urgent abdominal ultrasonographic assessment suggested the presence of an intestinal mass with a moderate amount of free fluid. A provisional diagnosis of an inflammatory appendicular mass was made. Ileocaecal tuberculosis and neoplasm were

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the other main differential diagnoses entertained. A decision for operative intervention was made.

At laparotomy, the mass was identified as a retrograde 25 cm long ileojejunal intussusception, 80 cm from the duodenojejunal flexure, which appeared congested but viable, resulting in an intraoperative decision to attempt reduction. Gentle reduction was successful, revealing a virtually gangrenous 20 cm segment of intussusceptum that was resected. Histopathological analysis showed a partially gangrenous thick-walled small-bowel segment with a pedunculated firm, partly necrosed polypoid structure measuring 3.0 × 2.5 × 2.5 cm macroscopically. Histology of this polyp revealed mainly connective tissue stroma with sparse vascularity and cellular content with no evidence of neoplasia, labelled a fibrous pseudopolyp. The patient recovered uneventfully. His routine follow-up review was unremarkable.

Case 2

A 47-year-old man presented to the outpatient department with a 4-month history of unexplained diarrhoea and periodic low-grade colicky abdominal pain associated with moderate nausea, which was erroneously diagnosed as persistent gastroenteritis and treated. On examination, he appeared well with no signs of anaemia. Abdominal and rectal examinations were unremarkable. All laboratory investigations including inflammatory, tumour and serological markers, including stool cultures, were normal.

At colonoscopy, an interesting pan-endoscopic real-time demonstration of intussusception was witnessed. The actual onset and progress of intussusception was revealed, showing the passage of a smooth lead-point polyp and the accompanying relayed intussusceptum coming through the ileocaecal valve into the caecum and progressing up to the hepatic flexure (Figure). This clinched the diagnosis immediately. The intussusception was reversible and reproducible after reduction by luminal insufflation followed by suction. Biopsies of the polyp only showed necrotic tissue.

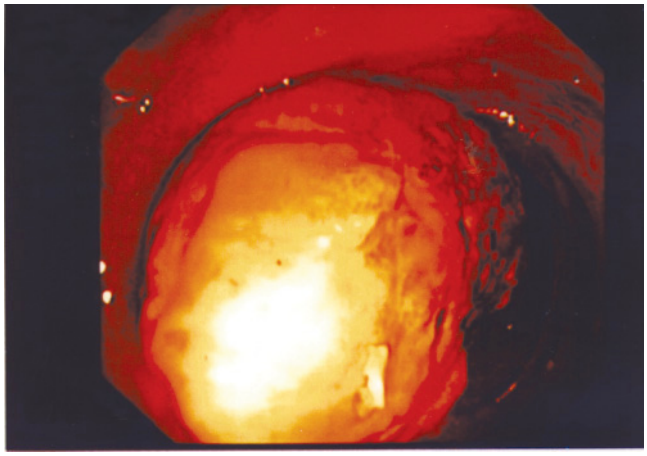

Figure. Endoscopic end-on view of intussusception. Note the appearance of the lead-point polyp.

At laparotomy, a terminal ileal polyp was easily palpable 18 cm from the ileocaecal valve. A limited right hemicolectomy was performed and a 30 cm segment of terminal ileum was removed. Two discrete mucosal polypoid lesions were observed within the terminal ileum. Histology of these polyps was similar, showing fibromyxoid connective tissue stroma containing numerous small vessels with a concentric arrangement of perivascular fibroblasts associated with eosinophils and lymphocytes. There was no evidence of malignancy. The patient recovered uneventfully and has remained well for 1 year following surgery.

Discussion

Adult intussusception is relatively rare, accounting for 1–3% of all surgically treated cases of adult bowel obstruction and 5–16% of all cases of intussusception per se.[1,7,8] It may be classified according to the site of occurrence on a descriptive anatomical basis, typically with the intussusceptum as the prefix (e.g. ileocolic, colocolic, enteroenteral, jejunogastric), by the direction of propulsion of the intussusceptum (antegrade vs retrograde) or by its aetiology (tumour-related, post-surgical, miscellaneous or idiopathic).[8] Virtually all are due to a definable intraluminal lesion, including surgically related mucosal changes secondary to an intestinal anastomosis, adhesions or post-gastrectomy; the idiopathic form is rare.[1,3,7,8]

Acute adult intussusception is uncommon compared with the chronic intermittent type.[1–3] In contrast, childhood intussusception tends to present acutely. Intussusception is rarely included in the differential diagnosis preoperatively, especially in the absence of a palpable mass (40% of cases).[1,3] Laparotomy is usually indicated for acute presentations.

A high index of suspicion is imperative for the more prevalent subacute intermittent intussusception, for these can produce few relevant results despite exhaustive investigations.[1,3] In the adult, these would simulate the far more commonplace symptoms of neoplastic growth. The most common presenting symptoms in this category of patients are abdominal pain (up to 90%), vomiting, rectal bleeding, melaena and anaemia.[2] Altered bowel habit or unexplained diarrhoea is uncommon.[1–3] It is, therefore, not surprising that misdiagnoses are common at presentation, with the most common differen-

tial diagnoses being abdominal mass of unknown aetiology, gastrointestinal bleeding and acute cholecystitis.[2]

Unlike the common ileocaecal childhood intussusception, the adult variety may occur in several sites. The commonest types are ileocolic and enteroenteral intussusception, which mirrors the higher incidence of small-bowel intraluminal entities as the causal factor (up to 65%). Although most small bowel pathologies are benign, there is a risk of malignancy of up to 50%, particularly of metastatic nodules and primary intestinal lymphoma.[2,3] In a recent series, up to one-third of intestinal lymphomas presented as intussusception noted at colonoscopy.[6] On the other hand, colonic intussusception must be assumed to harbour a malignancy at much higher rates of between 50% and 90%, with most of these being adenocarcinoma.[2,3,7]

Retrograde intussusception is rare and may occasionally be demonstrable endoscopically.[4] While ordinary antegrade intussusception is readily understood, particularly by virtue of the vector of intestinal peristalsis, retrograde intussusception defies any simple, physiologically based explanation. However, it is believed that the presence and degree of mobility of the lead-point polyp confers abnormal peristaltic activity to the immediate bowel.[1–3] This retrograde ball-valve effect may occur due to intermittent intussusception that causes the lead-point polyp to "button-hole" through an oedematous ring of more proximal mucosa, followed by its engulfment by the proximal intussicipiens. Alternatively, the preceding antegrade intussusception need never happen and retrograde intussusception may simply be caused by chance retrograde displacement of the tumour and abnormal peristalsis.[4]

Fibrous or fibroid pseudopolyps were implicated in both our cases. These are generally termed inflammatory fibroid polyps (IFPs), a recognized but rare pathological entity of unknown aetiology with unclear histopathological classification and pathogenesis.[9] This acquired condition of adulthood has several common characteristics. It is reportedly most common in the stomach, followed by the small bowel, and is only occasionally found in the colon.[9,10] IFPs can grow to be quite large, especially in the stomach.[10] IFPs arise from the submucosa and extensive infiltration may occur, which could be mistaken for gastrointestinal stromal tumours.[9,10] IFPs are usually discrete and singular, which made our finding in Case 2 of two synchronous ileal lesions relatively interesting, though this could be taken as a single afflicted segment due to an unknown common and localized inflammatory stimulus.

Characteristically, IFPs display a fibroblastic and vascular stroma with diffuse inflammatory eosinophilic infiltration and preferential immunohistochemical staining with vimentin, a non-specific marker for neuroectodermal tissue but also of fibroblasts and macrophages.[9] The presence of fibroblasts and the rich vascularity of the stroma are thought to be reactive. IFPs are not easily confirmed histologically by biopsy sampling alone: up to 90% of biopsies are non-confirmatory.[9,10] Only the smallest IFP can be removed by endoscopic snaring due to their apparent and inherent submucosal deep-seatedness.[10] IFPs are believed to have no malignant potential.[9,10] There is no direct association between IFPs and either tuberculosis or inflammatory bowel diseases.[9,11] IFPs have been shown to ulcerate and cause gastrointestinal bleeding and simple mechanical gastric and bowel obstruction as well as intussusception.[9,10]

Various radiological modalities have been used in the diagnosis of adult intussusception. The sensitivities of contrast studies such as barium enema and small-bowel follow-through are relatively poor.[1–3,7] Abdominal ultrasound lacks sensitivity and specificity. In our case, however, ultrasound provided enough information for the decision-making process. Abdominal computed tomography (CT) scan is the gold standard, with a 50–80% diagnostic rate demonstrating the supposedly typical "target" or "bullseye" sign, although this is not absolutely pathognomonic.[2,7] Nevertheless, no single radiological investigation should compromise operative intervention once a diagnosis of intussusception has been made in adults.[12]

Demonstration of intussusception at endoscopy is a chance finding and probably under-reported, and endoscopic therapy is limited.[4,6–8] Endoscopy is frequently neither safe nor feasible when intussusception is diagnosed and is not suitable oncological treatment for potentially malignant lead-point polyps.[6,10,13]

Bearing in mind the statistical risk of frank malignancy, the accepted wisdom for the treatment of adult intussusception is operative resection.[1–3] There are several areas of controversy here, including the question of intraoperative reduction; the extent of the resection and the approach taken for possible reduction and subsequent resection; and the role of laparoscopic surgery. Intraoperative reduction of an intussusception is controversial[1–3] and the possibility of potential tumour seeding of the peritoneal cavity must be taken into consideration. We believe that reduction has a place. It would seem sensible to perform selective reduction in viable cases of intussusception, especially of the small bowel, where the risk of overt malignancy is lower.[3] It is advisable for colonic intussusception to be left unreduced and resected as a single mass,

obeying standard oncological principles.[1–3] The extent of resection is a minor point of controversy. It is generally accepted that limited resection of small-bowel and ileum-based ileocolic intussusception is all that is necessary, because most of these polyps will be benign.

With current technological advances, laparoscopic surgery may have a role in the management of adult intussusception. The role of laparoscopic surgery in the literature has mainly been in manipulation and de-rotation of the associated volvulus, and especially in intracorporeal reduction.[14,15] Hand-assisted laparoscopic surgery may better facilitate these manoeuvres, as well as bowel mobilization, if required. Intra-abdominal resection may be simple or complex, ranging from the stapling of intussuscepting Meckel's diverticulum post-reduction to more complicated or even colonic cases; otherwise, it may be more prudent to perform extracorporeal resection via a minimal and convenient laparotomy access.[14,15]

The management strategy for acute situations is straightforward, with resuscitation and routine but urgent laboratory investigations. CT assessment may be considered but should not interfere with signs paramount to intestinal obstruction or peritonitis, for which laparotomy is mandatory. Management of subacute intussusception will rely on its chief presenting symptoms. As most investigations are non-specific, the most important tool is a high index of suspicion allied with CT imaging and colonoscopy. Colonoscopy is probably superior to contrast imaging for ileocolic intussusception, which carries a slightly higher risk of malignancy compared with more proximal enteroenteric types.[6] Nevertheless, the diagnostic algorithm for potential ileoileal intussusception is as yet unclear, with multimodality investigations prevailing. Once diagnosed, limited resection is advocated either at open surgery or through minimal-access surgery.

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